

Comparison of the efficacy of different treatment modalities for Kimura's disease

P. Ye¹, D.-Q. Ma¹, G.-Y. Yu¹, Y. Gao², X. Peng¹

¹Department of Oral and Maxillofacial Surgery, Peking University School and Hospital of Stomatology, Beijing, China; ²Department of Oral Pathology, Peking University School and Hospital of Stomatology, Beijing, China

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Abstract. The objective of this study was to investigate the clinical features of Kimura's disease in the head and neck region and to compare the local recurrence rate between three therapies used for the treatment of this disease. The clinicopathological information of 46 hospitalized patients suffering from Kimura's disease in the head and neck region over a 10-year period was reviewed retrospectively. All lesions were clinically observed in the head and neck region. These 46 patients underwent a total of 58 treatments; nine patients underwent multiple treatments due to local recurrence. Of the 58 treatments, 32 involved surgical excision alone, 24 involved surgical excision and postoperative low-dose radiotherapy (20–40 Gy), one was a combination of ultrasound-guided core needle biopsy and radiotherapy, and one was a combination of incisional biopsy and subsequent radiotherapy. During the follow-up period, nine patients suffered 16 local recurrences. The recurrence rate of surgical excision combined with low-dose radiotherapy was much lower than that of surgical excision alone or radiotherapy alone (both $P < 0.05$). It is concluded that Kimura's disease is a benign condition with a good prognosis, and surgical excision combined with postoperative low-dose radiotherapy is associated with the lowest local recurrence rate in the treatment of this disease.

Key words: Kimura's disease; recurrence; surgical excision; radiotherapy.

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In 1937, a Chinese oncologist named Kimm described seven patients with enlarged lymph nodes that exhibited pathological eosinophilic infiltration and termed this entity 'eosinophilic hyperplastic lymphogranuloma'.¹ Subsequently, Kimura et al. described the pathological features of this condition,² and since then Kimura's disease has become widely recognized.

Kimura's disease is a rare and chronic inflammatory lesion of unknown aetiology that most commonly presents in the head and neck region of Asian men. Clinical examination generally reveals subcutaneous swelling or nodules in the head and neck region that is partially associated with regional lymphadenopathy. The incidence peaks during the second and fourth

decades of life, and approximately 80–87% of patients are men.³ With a prolonged indolent course, Kimura's disease is known as a disfiguring condition, carrying no risk of malignant transformation even without treatment.

The definite aetiology and pathogenesis of Kimura's disease remains unclear. A recent study demonstrated that the

interaction between type 1 and type 2 helper T-cells may be involved in the development of Kimura's disease.⁴ Moreover, Day et al. suggested that Kimura's disease occasionally shows a clonal proliferation of T-cells.⁵ As Kimura's disease has an unknown aetiology, there is currently no preventative management for this condition.

No optimum treatment modality for symptomatic Kimura's disease is reported in the literature, although surgery, radiotherapy, steroid therapy, and intravenous immunoglobulin have proven effective.⁴ Surgical procedures are preferred for primary lesions without general contraindication, and postoperative steroid therapy is recommended, especially for patients with nephrotic syndrome.⁵ However, radiotherapy is considered satisfactory for recurrent cases or poor surgical candidates. Hareyama et al. recommended a dose of approximately 26–30 Gy for irradiation, and the radiation field should be confined to the area of the lesion and regional lymph nodes.⁶

The details of 46 patients with Kimura's disease are presented here. An analysis was performed of the patient clinical characteristics, with a focus on treatment regimens and corresponding outcomes. Furthermore, a comparison of the local recurrence rate of three therapies used for the treatment of Kimura's disease was conducted.

Patients and methods

Over a 10-year period (2004–2013), 46 patients with a clinicopathological diagnosis of Kimura's disease were treated at the study institution. Detailed clinical data were retrieved from the patients' medical charts, including sex, age, site involved, clinical duration, laboratory workup, treatment modality, and pathological diagnosis. Demographic information and details of these patients were analyzed. The clinical management of these cases was summarized, including the use of surgical excision, core needle biopsy, incisional biopsy, and low-dose radiotherapy.

Each treatment decision was made by a specific doctor based on their own experience. Surgical excisions were performed by different surgeons. As the salivary glands (submandibular gland, parotid gland, and minor salivary gland) were involved, the surrounding gland tissue was excised during surgery. Postoperative radiation of 20–40 Gy to a single field (2 Gy each day, 5 days a week) was delivered within 1 month after surgery.

The follow-up period ranged from 12 months to 120 months, with a mean period

of 65 months. Recurrence was evaluated through clinical and radiological examinations. The rate of loss to follow-up was 8.7%. Continued observation or further treatment for recurrent lesions was determined on the basis of the follow-up results.

For the statistical analysis, all data were analyzed using IBM SPSS Statistics version 20.0 software (IBM Corp., Armonk, NY, USA). The χ^2 test and Fisher's exact test were used to compare the recurrence rate between the different treatment modality groups; $P < 0.05$ was considered statistically significant.

Results

Clinical characteristics

Out of the 46 patients examined, 40 were male and six were female, giving a male-to-female ratio of 6.7:1. The age at onset ranged from 5 to 78 years (median age 41 years). With regard to the anatomical distribution, 23 cases (50%) involved the parotid region (six cases involved the bilateral parotid regions), 14 cases involved the submandibular region, and six cases involved multiple regions. Twenty-nine patients developed a solitary lesion of Kimura's disease. The remaining patients demonstrated multiple lesions to differing extents in the head and neck region upon clinical examination (Fig. 1).

Eight patients (17.4%) in this series presented with melanin pigmentation, five (10.9%) with coarseness, and nine (19.6%) with pruritus of the affected overlying skin. Spontaneous growth and decline was observed in two patients.

Only one patient presented with nephrotic syndrome; this patient died of subsequent renal failure. The laboratory

Table 1. Clinical features of 46 patients with Kimura's disease.

Clinical characteristics	No. of patients
Age at onset, years	
≤ 20	6 (13.0%)
20–40	16 (34.8%)
> 40	24 (52.2%)
Sex	
Male	40 (87.0%)
Female	6 (13.0%)
Anatomical location	
Parotid	23 (50%)
Submandibular	14 (30.4%)
Submental	1 (2.2%)
Cervical	2 (4.3%)
Multiple sites	6 (13.0%)
Peripheral blood eosinophilia	40 (87.0%)
Coarseness of overlying skin	5 (10.9%)
Pruritus	9 (19.6%)
Melanin pigmentation	8 (17.4%)
Spontaneous growth and decline	2 (4.3%)
Renal failure	1 (2.2%)

results of 40 patients (87.0%) revealed markedly elevated eosinophil counts in the peripheral blood (Table 1). Unfortunately, serum IgE levels were not recorded for this series.

The lesions most often appeared as hypoechoic masses or enlarged lymph nodes on preoperative ultrasonography. Typically, computed tomography revealed an expansive and ill-demarcated subcutaneous mass (Fig. 2). In addition, diffuse swelling of the nearby soft tissue could be seen in a few cases.

Management and prognosis

The 46 patients examined underwent 58 treatments; nine patients underwent



Fig. 1. Kimura's disease: the patient presented with bilateral parotid subcutaneous swelling, seen on frontal view.

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